

Case Report

Mixed Fungal Infection of Phaeohyphomycosis and Aspergillosis involving Right Maxillary Sinus in a Newly Diagnosed Case of Diabetes Mellitus

Nikita Modh¹, Shweta S. Joshi¹, Bhushan M. Warpe¹, Bhumika B. Prajapati^{1*}, Riddhi Yadav¹

¹Department of Pathology, Gujarat Adani Institute of Medical Sciences and G.K. General Hospital, Bhuj, Kachchh, Gujarat-370001

* Correspondence: Dr Bhumika B. Prajapati (prajapatibhumi7596@gmail.com)

ABSTRACT

Phaeohyphomycosis is an infection due to dematiaceous fungi which usually involve the skin and subcutaneous tissue. Phaeohyphomycosis in subcutaneous location has presentation in the form of nodules, papules ulcerated plaques, verruca-like hyperkeratotic plaques, cystic lesions, abscess formation, pyo-granuloma, discharging sinuses, non-healing ulcers. Brown-walled hyphae in the epidermis and dermis are its hallmark. Aspergillosis is ubiquitous, invasive fungal infection in diabetics and immunocompromised hosts. Here, we are reporting a mixed fungal infection of right maxillary sinus in a newly diagnosed case of diabetes mellitus

Keywords: Aspergillosis, Phaeohyphomycosis, Maxillary Sinus

INTRODUCTION

Phaeohyphomycosis involves heterogenous fungal organisms called “melanized” or “dematiaceous” fungi. Dematiaceous fungi are infected through inhalation. These fungi in their cell walls have melanin which imparts it the reliable stability in virulence.¹

Wide-spread Phaeohyphomycosis is an uncommon infection but its infectivity is on the rise in immunocompromised patients.² Numerous invasive presentations can be caused by these organisms including cerebral infection, pulmonary infection, deep local infections and disseminated disease. Rarely Phaeohyphomycosis can be seen with Aspergillosis, like in this case.

CASE REPORT

A 57 years old male presented with chief complaint of difficulty in breathing since past two months associated with on and off nasal discharge. It was not associated with sneezing, watering from eyes and

bleeding from nose. He had multiple visits at private practitioners for his complaints. He had family history of type II Diabetes. Relevant laboratory tests showed Hb as 14.4 g/dl, HIV test was non-reactive, HbA1c was 7.5%. Random blood sugar level (RBS) was 186 mg/dl.

CT scan of Para-nasal sinuses showed polyp like soft-tissue lesion involving right maxillary sinus causing widening and blockage of the same. It was suggestive of antro-choanal polyp which later became the clinical diagnosis.

Small multiple tissue bits were received as biopsy material in histopathology from right maxillary sinus. The multiple tissue bits were brown-black in colour; total measuring 1.8 cm x 1 cm x 0.4 cm and all bits were processed in one tissue block.

Microscopic examination: Hematoxylin and Eosin (H & E) sections showed fibro collagenous tissue lined by ciliated columnar to cuboidal epithelium. There was presence of fungal growth (Figure-1) having

pigmented ring-like structure called Globos with black yeast forms and pigmented hyphae along with balls of aspergillus fungus. Periodic Acid Schiff (PAS) stain (Figure-2) and Grocott–Gömöri's Methenamine Silver (GMS) stain (Figure-3) were used to confirm these fungal elements.

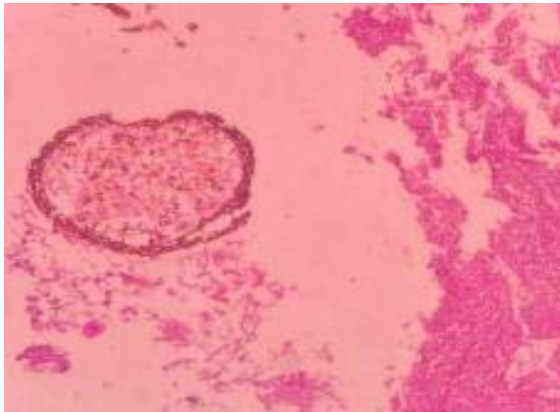


Figure-1: (H & E, X 100): Microphotograph of pigmented ring-like structure called Globos with black yeast forms and pigmented hyphae

DISCUSSION

The term “phaeohyphomycosis” was introduced by Ajello, et al¹ in 1974. It was done to specifically designate infections caused by filamentous, pigmented fungi that sustain melanin within the walls of their cells. It actually means “condition of fungi with dark hyphae.” “Dematiaceous” word comes from a Greek word meaning “bundle.” During the last few decades, phaeo-hyphomycosis has been linked to 1100 different species and 60 fungal genera. It has lead to many clinical syndromes, ranging from corneal inflammation and subcutaneous skin nodules to rapidly fatal, wide-spread systemic disease.¹⁻³

There are several mechanisms proposed by which melanin production by these fungi have virulence effect. It is believed to confer a protective advantage by removing free radicals with hypochlorites that are produced by phagocytic cells in their oxidative burst, which would normally kill most organisms.^{3,4}

Aspergillus fumigatus is a saprophytic fungus which is omnipresent in environment, especially in decaying

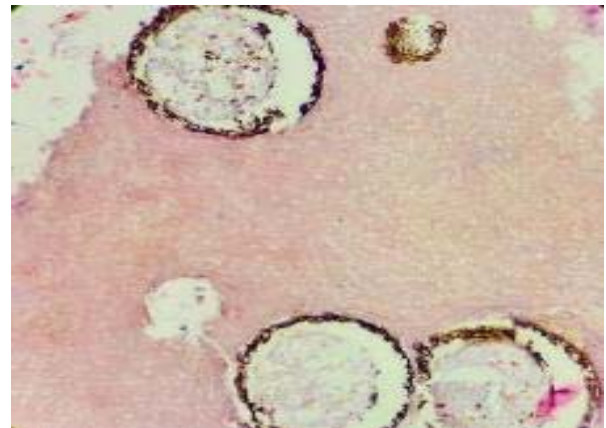


Figure-2: (PAS stain, X 100): Microphotograph of pigmented ring-like structure called Globos with black yeast forms and pigmented hyphae

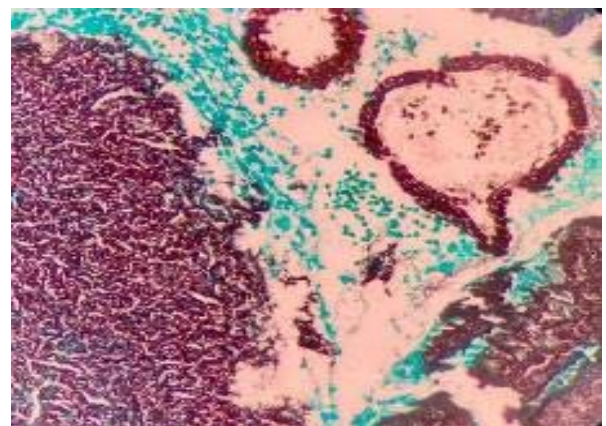


Figure-3: Microphotograph (GMS stain, X 100) of pigmented ring-like structures (Phaeohyphomycosis) and adjacent focus (left lower side) of filamentous fungi with acute angle branching (Aspergillosis)

vegetation. Conidia inhalation causes infection which is rapidly cleared in normal person but can lead to rapidly fatal, wide-spread systemic disease in immunocompromised hosts, individuals on long term steroid treatment and in diabetics. *Aspergillus fumigatus* dwells in environment in the optimal temperature of 37°C and at a pH range of 3.7 - 7.6.⁵

Damage caused by *Aspergillus fumigatus* can be due to fungal growth with invasion of nasal tissue or from inflammatory cellular recruitment at the infective nasal sites. Included in the latter are responses that are

ineffective in clearing the organism, occur in the process of immune reconstitution, or are associated with allergy. For example, in a murine model of chronic granulomatous disease, in which mice have defective phagocyte oxidase systems, administration of killed hyphae results in chronic inflammation due to persistence of fungal elements.⁶ Fungal stains like PAS, GMS are useful in highlighting the morphology. Hence, histopathology is a main diagnostic tool in the diagnosis of infective, subcutaneous skin nodules. FNAC can also be useful in some cases.⁷

Fungal culture was not done in our case due to clinico-radiological diagnosis of antro-choanal polyp and lack of suspicion of infective pathology. Cerebral and disseminated diseases are life-threatening and require improved therapeutic approaches, perhaps including combination of antifungal treatment and immunomodulation. These are key areas for future investigations.⁸

CONCLUSIONS

Freshly diagnosed case of type II diabetes mellitus can rarely present with mixed fungal infections involving the nasal cavity. As an eye-opener to clinicians, this case depicts the usage of special stains in biopsy and sending samples for microbiological studies like KOH mounting and Sabouraud dextrose agar (SDA) based culture study. Ideally samples for microbiological studies and histopathology/biopsy should be sent at the same time to laboratory in such infective cases.

Declaration of Patient Consent

We certify that we have obtained all appropriate patient consent forms. In the form the patient has given his consent for images and other clinical information to be reported in the journal. The patient understands that his names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

REFERENCES

1. Arcobello JT, Revankar SG. Phaeohyphomycosis. *Semin Respir Crit Care Med*. 2020;41(1):131-40.
2. Revankar SG, Patterson JE, Sutton DA, Pullen R, Rinaldi MG. Disseminated Phaeohyphomycosis:

Review of an Emerging Mycosis. *Clinical Infectious Diseases* 2002;34(4):467–76.

3. Revankar SG, Sutton DA. Melanized fungi in human disease. *Clin Microbiol Rev*. 2010;23(4):884-928.

4. Chhonkar A, Kataria D, Tambe S, Nayak CS. Three rare cases of cutaneous phaeohyphomycosis. *Indian J Plast Surg*. 2016;49(2):271-4.

5. Kwon-Chung KJ, Sugui JA. *Aspergillus fumigatus*-what makes the species a ubiquitous human fungal pathogen? *PLoS Pathog*. 2013;9(12):e1003743.

6. Hohl TM, Feldmesser M. *Aspergillus fumigatus*: principles of pathogenesis and host defense. *Eukaryot Cell*. 2007 Nov;6(11):1953-63.

7. Abraham LK, Joseph E, Thomas S, Matthai A. Subcutaneous phaeohyphomycosis: a clinicopathological study. *Int Surg J*. 2014;1(3):140-3.

8. Abdely HA. Phaeohyphomycosis: A Dark Question Mark in Clinical Disease. *Journal of invasive fungal infections*. 2009;3(3):82-8.

Source of support: Nil

Conflict of interest: None declared

How to cite:

Modh N, Joshi SS, Warpe BM, Prajapati BB, Yadav R. Mixed Fungal Infection of Phaeohyphomycosis and Aspergillosis involving Right Maxillary Sinus in a Newly Diagnosed Case of Diabetes Mellitus. *GAIMS J Med Sci* 2023;3(2):66-68. <https://doi.org/10.5281/zenodo.8135062>